

## REVIEW

# Special problems of children with myalgic encephalomyelitis/chronic fatigue syndrome and the enteroviral link

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*J Clin Pathol* 2006;**60**:125–128. doi: 10.1136/jcp.2006.042606

Since 1997, it has been known that myalgic encephalomyelitis/chronic fatigue syndrome constitutes the biggest cause of long-term sickness leading to absence from school, in both staff and pupils. The scale of the problem in children is substantial, and the pattern of illness in schools suggests a prominent role for viral infection—for example, the clustering of cases. The Dowsett–Colby study of 1997, researching long-term sickness, reported on a school roll of 333 024 pupils and 27 327 staff, and found a prevalence of long-term sickness in 70 of 100 000 pupils and 500 of 100 000 staff; 39% of cases were in clusters of three or more. The peak age was 14–16 years. The illness is known to be potentially severe and chronic. In addition, the Tymes Trust has reported that many affected children struggle for recognition of their needs, and are bullied by medical and educational professionals. Children should have time to recover sufficiently before returning to school; sustainable, energy-efficient and often home-based education is important here to fulfil legal obligations. Research is needed on viruses that trigger childhood myalgic encephalomyelitis—for example, enteroviruses—and on the neurocognitive defects caused by myalgic encephalomyelitis. We should recognise the value of previous biological research and records of outbreaks, and I recommend that myalgic encephalomyelitis be made notifiable owing to the encephalitic nature of the effects commonly reported in this illness.

biomedical study of ME/CFS in children, now in progress at the University of Dundee (Dundee, Scotland, UK), to ascertain whether the abnormalities found in small blood vessels in adult patients with ME/CFS, such as sensitivity to the endothelium-dependent vasodilator acetylcholine,<sup>3</sup> are present in children. Over the past 2 years, the Trust's professional guides for doctors and teachers have been placed on a free website at [www.tymes-trust.org](http://www.tymes-trust.org), which has been chosen for national archiving by the Wellcome Trust (London, UK). The Tymes Trust also runs a Professionals Referral Service (table 1), where medical, educational and social-service professionals can consult fellow professionals with experience in treating or educating children and young people with myalgic encephalomyelitis.

The Trust believes that it is important to remain true to the original aim of the children who started TYMES. It has always placed emphasis on the value of the patient's personal experience of the illness, now validated by the government's Expert Patient scheme. Thus, the Trust's Advice Line Team is unique in that all its members have personal experience of myalgic encephalomyelitis, particularly children, as well as the necessary professional guidance and information. In addition, the Tymes Trustcard pass card system (table 1), supported by the Association of School and College Leaders (Leicester, UK), is being used successfully in schools to support pupils when they are able to attend. The children's own voices are also published, including their activities, and information for parents and older readers, in *Vision* (table 1); is the only myalgic encephalomyelitis publication designed for an all-age market.

The emphasis of the government on the relevance of personal experience of chronic illness is one with which the Trust concurs; I value the combined approach of running the Trust and continuing to develop its initiatives despite residual limitations. It was only in 1985, when I myself was diagnosed with "royal free disease"<sup>4</sup> and then "myalgic encephalomyelitis", after a severe Coxsackie B viral infection, that I realised what having myalgic encephalomyelitis (the term used by the consultant microbiologist in my case) really meant. Nobody can prepare for such a dramatic and life-changing illness, not even someone who for many years has handled out-of-hours emergency calls in a busy urban area as I had done

Since 1997,<sup>1</sup> it has been known that myalgic encephalomyelitis constitutes the biggest cause of long-term sickness leading to absence from school, in both staff and pupils. The potentially severe and chronic disease, now called myalgic encephalomyelitis/chronic fatigue syndrome (ME/CFS) by the Department of Health,<sup>2</sup> isolates children and young people, who are often too ill to go out and need other ways of making contact with the world and of continuing their education.<sup>2</sup>

Young people themselves are often their own best advocates for this disease. As long ago as 1989, two young people with myalgic encephalomyelitis started their own newsletter, "TYMES" (The Young Myalgic Encephalomyelitis Sufferer). Their commendable initiative developed into a national charity, The Young Myalgic Encephalomyelitis Sufferers Trust ([www.tymes-trust.org](http://www.tymes-trust.org)), which has cofunded, along with ME Research UK and Search ME (Perth, UK), the first

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Accepted for publication  
3 August 2006  
Published Online First  
11 September 2006

**Abbreviations:** ME/CFS, myalgic encephalomyelitis/  
chronic fatigue syndrome

**Table 1** Special interest documents available at the Tymes Trust ([www.tymestrust.org](http://www.tymestrust.org))

Special interest document	Internet link to document
Professional referrals service	<a href="http://www.tymestrust.org/pdfs/referralservice.pdf">www.tymestrust.org/pdfs/referralservice.pdf</a>
The Tymes trustcard	<a href="http://www.tymestrust.org/pdfs/trustcardinfo.pdf">www.tymestrust.org/pdfs/trustcardinfo.pdf</a>
Vision	<a href="http://www.tymestrust.org/pdfs/vision2006-1.pdf">www.tymestrust.org/pdfs/vision2006-1.pdf</a>
Pushing the boundaries in ME/CFS*	<a href="http://www.tymestrust.org/pdfs/pushingtheboundaries.pdf">www.tymestrust.org/pdfs/pushingtheboundaries.pdf</a>
Back to school?*	<a href="http://www.tymestrust.org/pdfs/backtoschool.pdf">www.tymestrust.org/pdfs/backtoschool.pdf</a>
Succeeding with ME	<a href="http://www.tymestrust.org/pdfs/succeedingwithme.pdf">www.tymestrust.org/pdfs/succeedingwithme.pdf</a>
The SENCO's key role in supporting pupils with ME/CSF†	<a href="http://www.tymestrust.org/pdfs/senco.pdf">www.tymestrust.org/pdfs/senco.pdf</a>
GP's good practice guide to education for children with ME	<a href="http://www.tymestrust.org/pdfs/gpguide.pdf">www.tymestrust.org/pdfs/gpguide.pdf</a>
Ten points on the education of children with ME*	<a href="http://www.tymestrust.org/pdfs/tenpoints.pdf">www.tymestrust.org/pdfs/tenpoints.pdf</a>
Care of CFS/ME in children‡	<a href="http://www.tymestrust.org/pdfs/careinchildren.pdf">www.tymestrust.org/pdfs/careinchildren.pdf</a>

ME/CFS, myalgic encephalomyelitis/chronic fatigue syndrome; GP, general practitioner; ME, myalgic encephalomyelitis; SENCO, Special Educational Needs Coordinator.

\*First published in *Special Children* by Questions Publishing.

†First published in SENCO Update by the Birmingham University School of Education.

‡First published in *GP Magazine* by Haymarket Publishing.

for my husband's general practice. I had been awaiting the then housing Minister Ian Gow for a luncheon at the school where I was head teacher when violent pains in my back heralded what might have been influenza, but was not. My memory of the next few months is mostly blank. I was unable to rise from my bed, my body was continuously wracked with pain, I lost a good deal of my vocabulary, and when I did manage to reach the staircase, my brain had "forgotten" how to walk down it, and I fell. It was very similar to what I know of polio<sup>5-7</sup>; my breathing was affected, I could not grip anything and my legs refused to work. The pain continued, particularly in the large muscles of the back, stomach and thighs, which showed visible fasciculations, and also in the chest, where the pain was thought to originate in the intercostal muscles, and not in the heart itself, although I am not personally convinced of that. I was almost too weak to chew and, with, no curative treatment available; my disabilities were still so profound 5 years later, as is typical of myalgic encephalomyelitis,<sup>8</sup> that I officially left my profession to rebuild my health.

The scale of the problem in children and the pattern of illness in schools are of particular concern. There is clustering of cases,<sup>1-4</sup> often misunderstood as imitation of sick friends or siblings for secondary gain. Such gain is rare, with some children becoming at least as disabled as I was myself; children with less severe disease still lose their entire social lives, as relapse can occur if a person makes too much effort, which in some cases can be minimal.<sup>2</sup> The Dowsett-Colby study of 1997,<sup>1</sup> researching long-term sickness reports on a school roll of 333 024 pupils and 27 327 staff, found a prevalence of sickness in 70 of 100 000 pupils and 500 of 100 000 staff; 39% of cases were in clusters of three or more. The peak age was 14-16 years. When the BBC began to take an interest in misunderstandings surrounding the illness, it ran its own survey (BBC Panorama Survey, 1999; <http://news.bbc.co.uk/1/hi/events/panorama/506549.stm>), which I codesigned. There was a 62% response from 741 families surveyed, disclosing that 59% of families had been told that their child's illness was psychological; 15% of parents had been told that their own

psychological problems were the cause; 7% had been subject to child protection procedures; and 4% had been "labelled" as cases of Munchausen syndrome by proxy. None of these suspicions were to my knowledge upheld, and statistically they seem anomalous. It seemed that the children's illness was so severe for so long, and that both medical and educational professionals were finding it impossible to believe that these were physical impairments when the usual tests were inconclusive. We now have ample evidence of physical abnormalities in this illness, and I cite only a fraction of them.<sup>9-11</sup>

Myalgic encephalomyelitis has not uncommonly been mistaken for school phobia, anorexia nervosa, neglect, child abuse, Munchausen syndrome by proxy (fabricated or induced illness) or pervasive refusal syndrome. The common factor is often the perception of the illness as not capable of causing profound physical disability over a prolonged period, with cognitive-behavioural therapy mistakenly employed as a "cure". When an ME/CFS diagnosis is made, it is important to recognise that not only is the child's right to suitable education—that is, education in a manner suitable for their disabilities—enshrined in law, which includes education in the home where appropriate, but also this education must legally be provided after 15 days of illness to any child unable to access school.<sup>12</sup> In a study of 126 families carried out by The Young Myalgic Encephalomyelitis Sufferers Trust in 2003 (table 1), it was found that 87% had struggled for recognition of their needs and 81% had needed to move school to obtain recognition of their needs. Another concern was that 84% reported feeling bullied by the attitudes of professionals. Medical professionals can do a great deal to support the child's need for education in the home, whether it be by a teacher or by an interactive online system that encourages contact with other students (table 1).

The purpose of education is to educate; this may seem too obvious to state until we consider that attendance at school may be used as a measure of "recovery", whereas it is simply a measure of how often the child was able to get into school, whether or not this was educationally useful. Children with myalgic encephalomyelitis are not strong until they have substantially recovered. Intellectual performance seems to be impaired and reduced cortical blood flow has also been noted.<sup>13</sup> If the child has truly recovered, then the time spent in school should lead to an educational performance comparable to that of a healthy child. Often, it does not. According to one local education authority, the grades gained by children with myalgic encephalomyelitis using online education at home seem to be substantially higher than would normally be expected from children with myalgic encephalomyelitis studying through more conventional means (Warwickshire Local Educational Authority, 2005, personal communication). This may also be due to the typical pattern of intermittent attendance and absence that occurs when a child who is not robust has relapses due to attending school (table 1). Therefore, social considerations need to be balanced against legal rights to a suitable education to obtain qualifications. It is quite possible to provide for social contact separately from study, to gain the best of both worlds; children's health is thereby preserved, they can mix with friends when appropriate, and can also be proud of the qualifications they have achieved.

It is not commonly known that, although helpful and in some cases invaluable, there is no requirement for a consultant's recommendation for a child too unwell to access school to be provided with an alternative means of education. If there were, it is likely to be illegal, as few consultants could meet the 15-day statutory deadline. The recommendation by a general practitioner, as a qualified doctor, is sufficient. After a meeting with the Trust, Parliamentary Under-Secretary of State Lord Andrew Adonis wrote in a letter to The Young Myalgic

Encephalomyelitis Sufferers Trust: "It is unfortunate if, as calls to The Young Myalgic Encephalomyelitis Sufferers Trust advice line would appear to indicate, the advice in *Access to Education* is sometimes being misinterpreted as insisting that local authorities obtain a report from a consultant before action can be taken to provide support to children with myalgic encephalomyelitis who are unable to attend school full time. Our guidance is quite clear. We say that ideally an early diagnosis should be made by a consultant paediatrician but we do not suggest that this is a requirement."

As a former head teacher, I have spent many years developing special principles of education for children with myalgic encephalomyelitis that are working in practice (table 1). "Energy-efficient education" works by balancing the energy needed for healing against that needed for education. A child who is recovering is a child who can achieve educationally. Conversely, if the child becomes worse, education suffers. If educational methods make the child worse, then education has itself caused educational disadvantage. Education is something other than "schooling". We must set up a goal-oriented environment to help these children achieve what is not possible for them in an inappropriately structured one. We must also structure the curriculum appropriately, and make full use of special arrangements for academic examinations, including, if desirable, taking the examination at home to maximise the grade. Special arrangements are a legal right. The provisions set out by the Qualifications and Curriculum Authority and explained by the Trust do not aim to give preferential treatment to a child, but to level the playing field so that disadvantages in comparison with healthy children are, in so far as possible, minimised. Medical professionals are of invaluable support to children with myalgic encephalomyelitis as facilitators, recognising that achievement can occur only when the environment is structured to enable it to happen. Success is a huge morale booster. It helps sick young people to hold up their heads with pride.

One school of thought, which seems to treat myalgic encephalomyelitis as being perpetuated by psychological problems, says that children with myalgic encephalomyelitis need to be in school or they will not recover. The individual experience of families whom I advise as a professional educator indicates the opposite. This aspect of myalgic encephalomyelitis in children needs further study. Since the first article on myalgic encephalomyelitis in children and educational considerations was published in the *British Journal of Special Education*,<sup>14</sup> there has been a slow development in the recognition that children with myalgic encephalomyelitis need special educational provision, now enshrined in government statutory guidance,<sup>12</sup> and that this and taking into account not only the severity but also the stage of the illness helps improve health.

Now, there is a pressing need to consider developing our epidemiological knowledge of myalgic encephalomyelitis. The late consultant paediatrician Dr Alan Franklin had a maxim: "If the child doesn't fit the theory, then the theory is wrong". Suspicions among some doctors that groups of children with myalgic encephalomyelitis are copying one another, rather than being actually physically ill, have led not only to misunderstandings and obstacles to the special educational needs provision but also to the sidelining of an epidemiological phenomenon: children may well be the epicentre of this illness. The Dowsett-Colby study<sup>1</sup> was conducted to determine "whether the recognition of multiple cases of ME/CFS in one school is an [a] unique experience". In the 1098 schools situated in six English local education authority areas that were investigated, the diagnosis of ME/CFS was markedly associated with case clustering. The outbreak of "summer flu" with

gastroenteritis in a village school, which had prompted the study, had affected 70 of 230 children aged 9–11 years. In the autumn term, about 10% of those affected in the summer experienced profound fatigue and cognitive disturbance that had not resolved on follow-up 2 years later. One child's encephalitic illness was shown to be due to an enterovirus infection in July, but investigations in other cases were delayed and were therefore unhelpful. Relapse in the same pupils occurred the next year.

The Trust has proposed four priorities for research, giving evidence at the Gibson Parliamentary Inquiry into myalgic encephalomyelitis. These are as follows:

**Priority 1:** That an enteroviral study be set up as suggested by Chia<sup>15</sup> in his review of the role of enterovirus in chronic fatigue syndrome.

Dr Chia explains that although "initial reports of chronic enteroviral infections causing debilitating symptoms in patients with CFS were met with scepticism" and had been "largely forgotten" for the past decade, "observations from in vitro experiments and from animal models have clearly established a state of chronic persistence through the formation of double-stranded RNA, similar to findings reported in muscle biopsies of patients with CFS". He goes on to say that recent evidence has not only confirmed the earlier studies but also clarified the pathogenic role of viral RNA through antiviral treatment.

Dr Chia summarises the available experimental and clinical evidence that supports the role of enterovirus—the virus family that includes the polioviruses. He quotes tests showing that concentrations of antibodies against enteroviruses in patients with CFS were still raised >48 months after the acute phase. Pointing out methodological flaws with studies that once threw doubt on the enteroviral connection, he concludes:

Thus, renewed interest is needed to study further the role of enterovirus as the causative agent of CFS. [...] A well designed, randomised, controlled trial of antiviral treatment will ultimately provide crucial information on the pathogenic role of enterovirus in patients with CFS and other chronic diseases.

**Priority 2:** That ME/CFS be made notifiable in schools—staff as well as pupils—so as to show the pattern in school-age children (these include mini-epidemics and clusters).

Encephalitis is already notifiable. In adopting the term chronic fatigue syndrome, we have blurred the distinction between those with the encephalitic condition known as myalgic encephalomyelitis as described by Dowsett *et al*<sup>8</sup> and those who have a more generalised condition.

**Priority 3:** That all previous work on myalgic encephalomyelitis epidemics over the decades—for example, during the whole of the 20th century—be revisited; it has been sidelined in the adoption of the unsatisfactory name chronic fatigue syndrome.

Many of the epidemics were in schools and this knowledge is vital to revisit and correlate with our increased knowledge of virology in the 21st century.

**Priority 4:** That there be further studies on cognitive impairments caused by ME/CFS.

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Competing interests: None declared.

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